Urological Science 23 (2012) 93–95

Contents lists available at SciVerse ScienceDirect

Urological Science

journal homepage: www.urol-sci.com

Case report

Giant suprarenal tumor

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ARTICLE INFO

Article history:
Received 9 December 2010
Received in revised form
1 March 2011
Accepted 23 March 2011
Available online 13 August 2012

Keywords:
Hounsfield units
suprarenal tumor

ABSTRACT

Adrenal tumors evoke considerable interest and represent various diagnostic challenges. Adrenal tumors can be stratified into adrenal medullary and adrenocortical tumors. Approximately 60% of adrenocortical tumors are hormonally active and show specific signs and symptoms. Patients with a nonfunctioning adrenal tumor usually present with abdominal discomfort due to the mass effect of the tumor. An imaging feature that differentiates benign from malignant adrenal neoplasms is the tumor size. Thus, for the differential diagnosis of adrenal lesions, measurement of the Hounsfield units on an unenhanced computed tomographic scan is of great value when differentiating malignant from benign lesions. Herein, we describe a young female patient who presented with a huge left suprarenal tumor. She underwent complete resection of the adrenal tumor. The final pathological diagnosis was an adrenocortical carcinoma. There has been no evidence of recurrence for the last 4 years.

1. Introduction

Suprarenal tumors evoke considerable interest and represent various diagnostic challenges to physicians. Adrenal tumors consists the most portion of suprarenal tumors, which can be stratified into adrenal medullary and adrenocortical tumors. Approximately 60% of adrenocortical tumors are hormonally active and show specific signs and symptoms. Patients with a nonfunctioning adrenal tumor usually present with abdominal discomfort due to the mass effect of the tumor. An imaging feature that differentiates benign from malignant adrenal neoplasms is the tumor size. Based on the report of this case and subsequent review of the literatures, measurement of the Hounsfield units on an unenhanced computed tomographic scan is of great value when differentiating malignant from benign lesions.

2. Case report

A 36-year-old woman was referred from the nephrology clinic due to the presence of a palpable mass in her left loin region. She denied any symptoms such as abdominal pain, face flushing, headaches, weight loss, or any other systemic symptoms. There was also no contributory medical history. A physical examination revealed the presence of a palpable mass over the upper left quadrant of the abdomen that extended up to the flank region; however, the mass lesion was not found to be tender. Results of abdominal computed tomography (CT) scan revealed a huge left suprarenal mass. There were evidences of calcification and central necrosis within the tumor, which had also downwardly displaced the left kidney (Figs. 1A and 1B). All laboratory tests were normal, including urinalysis, complete blood count, and serum chemical studies. An adrenal endocrine survey, including cortisol, testosterone, aldosterone, progesterone, and catecholamine, was also within normal limits. Metastatic evaluation, including a whole-body bone scan and positron emission tomography (PET), revealed a hot spot in the right temporal bone. However, there was no bony lesion on the plain skull film.

2.1. Differential diagnosis

Abdominal CT scan after contrast medium enhancement revealed an inhomogeneous mass with irregular enhancement of the solid components. Calcifications and central necrosis were also visible. No surrounding tissues or organs were involved. The possible differential diagnoses were adrenal medullary-type tumors (pheochromocytomas and rare ganglioneuromas), adrenocortical tumors [adenomas, myelolipomas, oncocytic neoplasms, and adrenocortical carcinomas (ACCs)], neurogenic tumors (such as

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http://dx.doi.org/10.1016/j.urols.2012.07.002

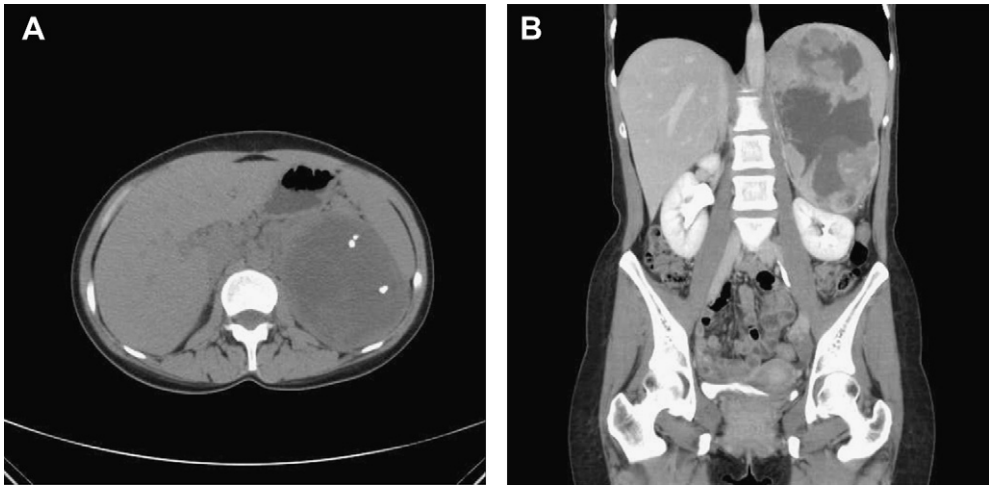

Fig. 1. (A) Noncontrast unenhanced computed tomographic (CT) scan revealing a well-defined huge mass with severe necrosis and evident calcifications. (B) Contrast-enhanced CT, showing a delayed attenuation of approximately 56–78 HU (on an approximately 10–15-minute delayed enhanced CT).

schwannomas), and retroperitoneal sarcomas (malignant fibrohistiocytomas and leiomyosarcomas). As is the case with any mass involving the adrenal glands, it is important to differentiate the lesion from an adrenal adenoma. An imaging feature that differentiates benign from malignant adrenal neoplasms is the tumor size.[1] A diameter of <5 cm usually suggests a benign origin. Furthermore, measurement of the Hounsfield units on an unenhanced CT scan is of great value when differentiating malignant from benign lesions. An attenuation value of >10 HU on an unenhanced CT scan or an enhancement washout of <50% and delayed attenuation of >35 HU (on approximately 10–15-minute delayed enhanced CT scan) suggests a malignancy.[2] This young female patient had a giant nonfunctional suprarenal tumor that gave attenuation values of approximately 26–35 HU on the unenhanced CT scans and values of approximately 56–78 HU on enhanced CT scans, and therefore, an ACC was suspected. However, it was not possible to rule out the rare possibility of an oncocytic neoplasm, because a pathological feature that is useful when identifying an adrenal oncocytic tumor is a fibrous capsule.[3]

2.2. Management

Considering the huge size of the mass and the potential possibility of malignancy, the patient underwent open surgery via a transabdominal approach using a subcostal incision; this facilitated maximal exposure for the complete surgical excision of the tumor and also minimized tumor spillage. An ovoid, well-circumscribed tumor (16 × 9 × 8.5 cm) distinct from the left kidney was found. The tumor had compressed the spleen and pancreas and stretched the descending colon, but had not invaded these organs. The tumor was completely removed. The patient was discharged on the 6th day after the operation. No adjuvant chemotherapeutic drug such as mitotane was administered. The patient has had regular follow-ups including chest X-rays and CT scans every 3 months and a yearly PET scan. There has been no evidence of recurrence over the last 4 years.

2.3. Pathology

The final pathological diagnosis was an ACC. Upon sectioning, we found that the encapsulated tumor was a heterogeneous mass with a brownish-pink to yellow color on the cut surface and showed multiple areas of myxoid change and necrosis. The surgical margin was free of tumor. A microscopic examination revealed a mass composed of round to polygonal neoplastic cells with abundant eosinophilic cytoplasm (Fig. 2A). These nuclei were hyperchromatic and pleomorphic, and had obvious nucleoli and mitotic activity [2 per 50 high power fields (HPFs)]. Tumor necrosis was evident. Neither capsular nor vascular invasion was identified. The pathological stage was T2N0M0 (European Network for the Study of Adrenal Tumor, the ENSAT staging system). Immunohistochemical analysis revealed that the tumor cells were strongly positive for cytokeratin (Fig. 2B) and desmin, but were weakly positive for vimentin and S-100 protein, and negative for Ki67 (Fig. 2C).

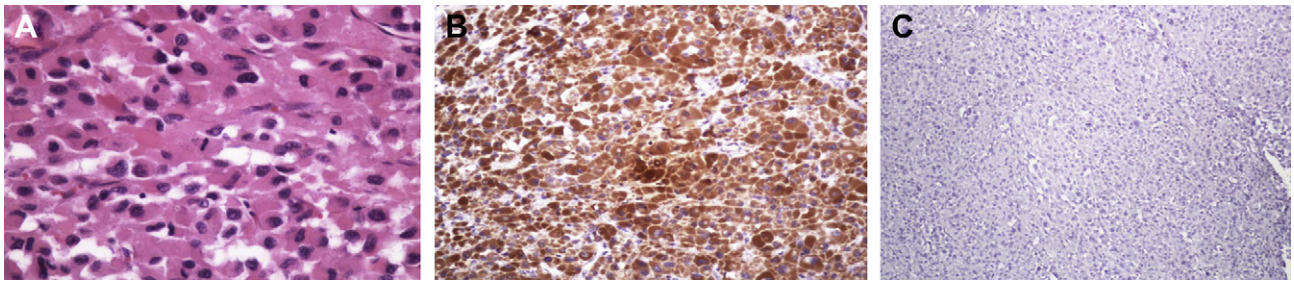

Fig. 2. (A) Microscopic analysis revealed the presence of obvious nucleoli, mitotic activity, and eosinophilic tumor cell cytoplasm when the sections were stained with hematoxylin and eosin stain (400×). (B) Immunohistochemical analysis revealed that the tumor cells were strongly positive for cytokeratin (200×) and (C) negative for Ki67 (100×).

3. Discussion

All incidentally discovered adrenal masses should be checked for excess hormonal secretions, including cortisol, aldosterone, and catecholamine. The most common complaint associated with nonfunctional adrenal tumors is abdominal pain and/or a palpable tumor. The most common first-line imaging modality is a CT scan. Typical radiographic features associated with a malignant adrenal tumor are a large size (>5 cm), vague borders, an irregular shape, and a heterogeneous texture. However, in the end, ACC is a histological diagnosis, and a pathological diagnosis may be difficult due to the lack of clear-cut morphological criteria. The Weiss score is most widely used when grading the microscopic features that are suggestive of a malignant tumor. The classification includes nine histological criteria which can predict adrenocortical tumors that metastasize or recur. We noticed a high nuclear grade (Fuhrman grade: approximately 3–4), a high mitotic rate (>5/50 HPF), atypical mitotic figures, eosinophilic tumor cell cytoplasm, a diffuse architecture present in ≥33% of the tumor, necrosis, invasion of venous structures, invasion of sinusoidal structures, and capsular invasion.[4] Immunohistochemical stains such as Ki67 can be used for both differentiating benign from malignant tumors and determining the prognosis of ACC. A Ki67 index of >7% is associated with significantly shortened disease-free survival.[5]

The treatment of choice and the only chance to cure ACC is complete resection of the tumor, and if necessary, an adjacent lymphadenectomy is mandatory for a cure. The anterior abdominal approach offers easy access to the main vessels and good exposure that prevents tumor spillage when resecting larger tumors. The long-term outcome did not significantly differ between the use of a laparoscopic adrenalectomy and open surgery for tumors of <10 cm.[6] Adjuvant therapy is recommended for most patients because of the risk of recurrence, which is high at approximately 60–80%.[7] Mitotane can be administered to treat inoperable tumors and metastatic disease or when there is incomplete resection. Radiation therapy of the tumor bed is recommended for patients with a histologically incomplete (R1) or undetermined (Rx) resection. Based on the tumor-free resection margin and the negative immunostaining for Ki67, adjuvant treatment was not suggested to this patient.

An ACC is a rare malignancy, and a multidisciplinary approach has the best chance to provide optimized management of this lethal orphan disease. An improved understanding of the pathogenesis of this malignancy and a collaborative effort will lead to therapeutic advances that may improve treatment outcomes.

Conflicts of interest statement

The authors declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

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